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LETTERS TO THE EDITOR

Cost-Effectiveness of Expanded Newborn Screening in Texas

We read with great interest the study by Tiwana et al. [1] regarding a critical issue in public health: cost-effective measures in expanded newborn screening. According to their results, although expanded screening was associated with increased costs, it improved quality of life. Therefore, the authors concluded that expanded newborn screening, as compared with unexpanded screening, was a cost-effective option in Texas. Some analytic problems in this article, however, need further clarification.

The authors likely miscalculated the incremental cost-effectiveness ratio (ICER) in Table 6. Conventionally, the ICER is calculated by using the following formula:

$$\text{ICER} = (\text{Cost}_{\text{with screening}} - \text{Cost}_{\text{without screening}})/(\text{Effect}_{\text{with screening}} - \text{Effect}_{\text{without screening}})$$

While in Table 6, the ICER was miscalculated as

$$\text{ICER} = [(\text{Cost}_{\text{with screening}} - \text{Cost}_{\text{without screening}})/\text{Effect}_{\text{with screening}}] - \text{Effect}_{\text{without screening}}$$

The corrected Table 6 content is listed below. Based on our corrections, the ICER should be $47,079 per quality-adjusted life-year (QALY) for arginosuccinic acidemia and citrullinemia, $5,254 per QALY for maple syrup urine disease, $3,397 per QALY for medium chain acyl-CoA dehydrogenase deficiency (MCADD), $15,378 per QALY for glutaric acidemia type I, and $4,973 per QALY for classical organic acid disorders. Therefore, it would be better that the relevant parts in the main text, including the first paragraph of the Results (p. 616) and the second paragraph in the Discussion (p. 618), are carefully reexamined and properly readdressed.

Original

Table 6 – Average cost and effectiveness by disorder at 3%

	With screening		Without screening		ICER ($/QALY)
	Cost ($)	Effectiveness	Cost ($)	Effectiveness	
ASA_CIT	681,455.00	14.34	538,334.00	11.3	9,969.24
HCY	267,699.00	24.09	333,594.00	21.04	Dominant
MSUD	136,607.00	25.03	36,303.00	5.94	4,001.41
MCADD	266,711.00	24.56	250,678.00	19.84	632.97
GA-I	291,269.00	24.73	217,145.00	19.91	2,977.42
COAD	184,436.00	23.46	142,468.00	15.02	1,773.90

ASA, arginosuccinic acidemia; CIT, citrullinemia; COAD, classical organic acid disorders; GA-1, glutaric acidemia type I; HCY, homocystinuria; ICER, incremental cost-effectiveness ratio; MCADD, medium chain acyl-CoA dehydrogenase deficiency; MSUD, maple syrup urine disease; QALY, quality-adjusted life-year.

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Corrected

Table 6 – Average cost and effectiveness by disorder at 3%

Disorder	With screening		Without screening		ICER ($/QALY)
	Cost ($)	Effectiveness	Cost ($)	Effectiveness	
ASA_CIT	681,455.00	14.34	538,334.00	11.3	47,079.28
HCY	267,699.00	24.09	333,594.00	21.04	Dominant
MSUD	136,607.00	25.03	36,303.00	5.94	5,254.27
MCADD	266,711.00	24.56	250,678.00	19.84	3,396.82
GA-I	291,269.00	24.73	217,145.00	19.91	15,378.42
COAD	184,436.00	23.46	142,468.00	15.02	4,972.51

ASA, arginosuccinic acidemia; CIT, citrullinemia; COAD, classical organic acid disorders; GA-1, glutaric acidemia type I; HCY, homocystinuria; ICER, incremental cost-effectiveness ratio; MCADD, medium chain acyl-CoA dehydrogenase deficiency; MSUD, maple syrup urine disease; QALY, quality-adjusted life-year.

The corrected ICER is more consistent with that in previous studies. Insinga et al. [2] reported an estimate of $41,862 per QALY for MCADD in Wisconsin. Prosser et al.'s [3] study disclosed that the cost-effectiveness of newborn screening for MCADD was $27,423 per QALY. In Canada, Cipriano et al. [4], who assessed the cost-effectiveness of expanding newborn screening for six diseases, showed that the ICER was $1,440,777 per life-year (LY) gained for arginosuccinic acidemia, $1,753,719 per LY gained for citrullinemia, $331,200 per LY gained for homocystinuria, $15,426 per LY gained for maple syrup urine disease, $62,798 per LY gained for MCADD, and $48,071 per LY gained for glutaric acidemia type I [4].

Although the threshold of cost-effectiveness for medical interventions is roughly £20,000 to 30,000 in the United Kingdom, and US $50,000 to $100,000 in the United States, it is well accepted that these values are unjustified [5]. The use of $50,000 as a limit to determine cost-effectiveness was set in the early 1980s, and was used widely after 1996 [6]. Many recent studies have challenged these values by using willingness to pay [5–7]. For example, the willingness-to-pay values in Shiroiwa et al.'s [5] study were £23,000 in the United Kingdom and US $62,000 in the United States.

Despite the unjustified cost-effectiveness limit, the ICER of expanded newborn screening in this study, $11,560 per QALY, is still far below the limit. The conclusion that expanded newborn screening is a cost-effective strategy, despite the errata mentioned above, remains robust and convincible.

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